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Urological Science

journal homepage: www.urol-sci.com

Case report

Penile-preserving surgery for primary urothelial carcinoma of male urethra

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ARTICLE INFO

Article history:

Received 7 April 2014

Received in revised form

26 February 2015

Accepted 2 March 2015

Available online 10 April 2015

Keywords:

primary urethral carcinoma

transurethral resection

urothelial carcinoma

ABSTRACT

Primary urethral carcinoma is a rare cancer, comprising <1% of all malignancies. The location of this lesion presents a certain dilemma of treatment between efficacy and quality of life. We report an 84-year-old male patient, with a history of chronic hepatitis C, hypertension, and transient ischemic accident, who presented with dysuria and acute urinary retention. The intravenous urography showed mild prostatic enlargement, but no stone or filling defect was noted in the upper urinary tract. On urethroscopy, multiple papillary tumors were found at the pendulous urethra, and the pathology of biopsy confirmed urothelial carcinoma. The patient was admitted, and electroresection with fulguration of urethral tumors was performed owing to the patient's old age and poor performance status. Intra-urethral and intravesical chemotherapy with mitomycin C was regularly given at the outpatient clinic. Recurrent urothelial carcinomas were noted twice in the first 2 years of follow up, and repeated transurethral resections were done. Unfortunately, liver cirrhosis with hepatocellular carcinoma was diagnosed last June, for which he received transcatheter arterial chemoembolization. No recurrence of urethral cancer has been found on semiannual cystoscopy in the past 3 years. Penile-preserving surgery is a reasonable surgical option for elderly primary urethral carcinoma patients with acceptable oncological outcome and good quality of life.

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1. Introduction

Primary urethral carcinoma (PUC) is a rare malignancy, comprising <1% of all malignancies, and having an incidence rate of 4.3/million in males and 1.5/million in females.¹ Risk factors include urethral stricture, chronic inflammation or irritation of urethra, and irradiation therapy. Histologic types are mainly transitional cell carcinoma (55%), squamous cell carcinoma (21.5%), and adenocarcinoma (16.4%).¹ The overall survival and disease-specific survival rates at 5 years were 42% and 50%, respectively.²

The treatment of localized PUC in males has followed the procedure for penile cancer with aggressive surgical excision in the past. According to the European Association of Urology 2013 guidelines on PUC, penile-preserving surgery could now be deemed as an alternative to primary urethectomy.³ Age, grade,

TNM stage, histology, and extent of surgery were prognostic factors predictive of overall and cancer-specific survival.⁴

We report a case of primary urothelial carcinoma of distal urethra in an 84-year-old man, who was treated with electroresection with fulguration and followed up at our urology clinic for 4.5 years.

2. Case Report

An 84-year-old man visited our urology clinic with a chief complaint of voiding difficulty for 2 weeks in December 2009. He had a history of chronic hepatitis C, hypertension, transient ischemic accident, and cigarette smoking for >30 years. At the clinic, digital rectal examination revealed a walnut-sized prostate with a rubbery consistency and smooth surface. No palpable or visible lesion was observed in the external genitalia or inguinal area on physical examination. The intravenous urography showed mild basal indentation of the urinary bladder due to benign prostatic hyperplasia. No filling defect was noted in the urinary bladder or the upper urinary tract (Fig. 1A).

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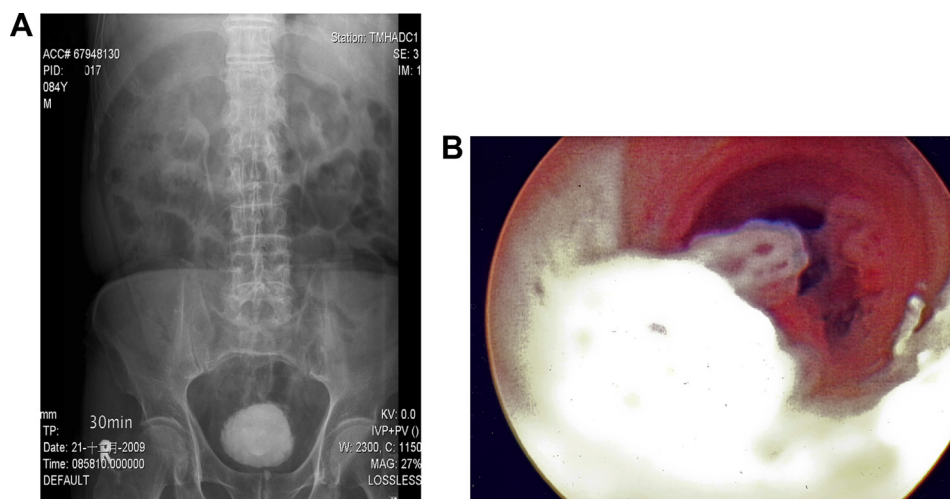


Fig. 1. (A) Intravenous urography shows mild basal indentation of the bladder owing to benign prostatic hyperplasia (BPH); patent bilateral ureters and no filling defect in kidney and urinary bladder. (B) Urethrocystoscopy reveals multiple papillary tumors of the pendulous urethra.

His complete blood cell count and blood biochemistry were normal except for an elevated liver function test result (aspartate aminotransferase 74 IU/L, and alanine transaminase 64 IU/L). The prostate-specific antigen level was 4.1 ng/dL, which was normal for his age. Urinalysis revealed mild pyuria (5–10 white blood cells/high power field). Urethrocystoscopy was arranged, and multiple papillary tumors at the pendulous urethra were found 7–8 cm from the urethral meatus (Fig. 1B). Cystoscopic cold-cup biopsies were done, and the pathology report confirmed grade 1 urothelial carcinoma of the urethra at T1 stage (limited at lamina propria; Fig. 2). An abdominal computed tomography was performed, which showed no lymphadenopathy or distant organ metastasis.

The patient was admitted for surgical intervention, and electroresection of the urethral tumor with fulguration was performed because of his (old) age. He was discharged home with medication for benign prostatic hyperplasia. Intraurethral and intravesical chemotherapy with mitomycin C (30 mg) was administered at our urology clinic weekly for 4 weeks and then monthly. Mitomycin C dissolved in 30 mL of normal saline was instilled along the way from the urinary bladder to the distal urethra by slowly pulling out the urethral catheter. A penile clamp was applied to the distal urethra for 30 minutes after instillation. Two recurrences of PUC were noted on follow-up urethrocystoscopy in March 2010 and July 2012, respectively. Repeated transurethral resections of urethral tumors with fulguration were performed. Urethral stricture with

urinary retention occurred once in October 2013 and was relieved by optic urethrotomy.

The patient also had chronic hepatitis C-related liver cirrhosis, Child-Pugh B, with ascites and hepatocellular carcinoma, T2N0M0. Transcatheter arterial chemoembolization was performed for the hepatocellular carcinoma in June 2013. No recurrence of urothelial carcinoma of the urethra has been noted at follow up in the past 2 years.

3. Discussion

PUC is an uncommon cancer and can metastasize to tissues around the urethra and inguinal or pelvic lymph nodes. The rarity and location of PUC render its treatment option a difficult choice between efficacy of tumor control and quality of life. Therefore, there is a lack of consensus on the optimal treatment for PUC. There is an agreement that clinical management should be primarily based on the clinical stage and location of the tumors. The overall survival rate was 83% for superficial disease versus 36% for more advanced cancers.² Clinical staging provides the strongest prognostic indication of survival.⁵ Early detection affords the best outcome of treatment.

Symptoms and signs of PUC include hematuria, urethral bleeding, urethral discharge, dysuria, frequent urination, a lump in the penis, and enlarged lymph nodes in the inguinal area. The

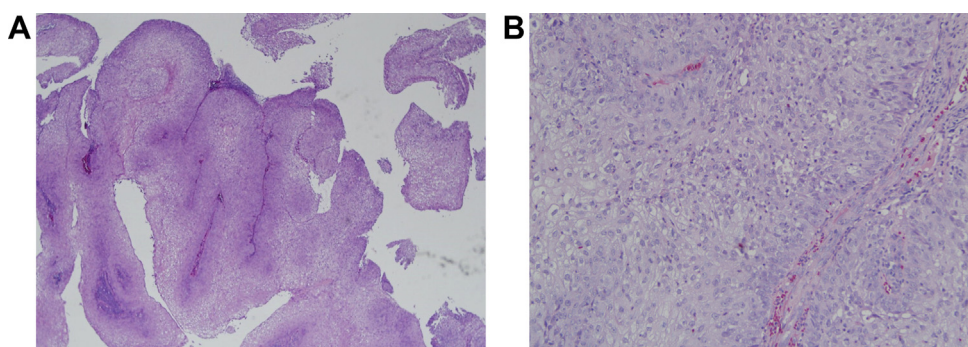


Fig. 2. (A) Histologic examination of the cystoscopic biopsy of the urethral tumor shows low-grade urothelial carcinoma with papillary growth pattern, branching vascular stalks, and increased cell layers (hematoxylin–eosin stain, 20×). (B) The tumor cells show relatively uniform nuclei with some prominent nucleoli and common mitosis (hematoxylin–eosin stain, 400×).

overall survival rate was 26% for bulbar urethral cancer and 69% for anterior urethral cancer.² It is pivotal to find and locate the urethral tumor at early stages with cystoscopy. Intravenous urography and computed tomography scans are also important tools for checkup of the upper urinary tract and pelvic and abdominal lymph node metastases. Magnetic resonance imaging can provide more local anatomic detail and excellent soft-tissue contrast that can increase the accuracy of staging, so it may serve as a better alternative for image checkup.

Most of the information of treatment of PUC is derived from retrospective and single-center case series. The choice of treatment is hence not standardized owing to the rarity of PUC. Surgery remains the cornerstone of therapy, and the approach depends on tumor grade, stage, and location. Previously, radical surgery to achieve a wide safety margin was recommended. The European Association of Urology has issued guidelines on PUC in 2013 and recommended penile-preserving surgery as an alternative to primary urethrectomy, if negative surgical margins can be achieved.³ Ablative surgical techniques, such as electroresection with fulguration, or laser vaporization with coagulation, can be used to preserve the penis and urethra in aged patients with superficial distal urethral cancer.⁶

Invasive primary carcinoma of the male urethra has a dismal outcome even with extensive surgical extirpation or radiation therapy. Coordinated chemoradiation therapy has been postulated for the primary treatment of invasive PUC with good response. The 5-year overall and disease-specific survival rates were 60% and 83%, respectively.⁷ Multimodal treatment with surgery and chemoradiation is the optimal treatment for advanced urethral carcinoma.

In our case, this 84-year-old patient had received three transurethral resections, and no recurrence has been noted in the past 2

years. It is consistent with the recommendation in the literature that superficial distal urethral cancer could be treated with endoscopic transurethral resection with preservation of organ function.

In conclusion, PUC has a more favorable prognosis in early stage and anterior urethra. Superficial anterior urethral carcinoma can be managed endoscopically using electroresection or laser vaporization, especially in aged patients, who are not legitimate candidates for radical surgery.

Conflicts of interest

All contributing authors declare no conflicts of interest.

Sources of Funding

No funding was received for the work described in the article.

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